European Journal of Paediatric Neurology 41 (2022) 36–40

Contents lists available at ScienceDirect

European Journal of Paediatric Neurology

journal homepage: www.journals.elsevier.com/european-journal-of-paediatric-neurology

Dystonia during hand activity in children with spastic unilateral cerebral palsy, an observational study

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ARTICLE INFO

Keywords:
Dyskinesia
Screening
Spastic unilateral cerebral palsy
Upper extremity
Dystonia

ABSTRACT

Background: Spasticity and dyskinesia are motor signs that co-exist in cerebral palsy (CP). It is well accepted that, in spastic bilateral CP, dystonia can be present in addition to spasticity, and equally that spasticity is often present in individuals with dyskinetic CP. In unilateral spastic CP, dystonia of the upper extremity is only rarely identified or addressed. The aim of this study was to investigate if dystonia was present in the hand of children with unilateral spastic CP, and, if present, to what extent, and when it was first noticeable.
Method: Ninety-seven children with unilateral spastic CP, born 1999–2014, with standardized digital films of hand function from Assisting Hand Assessments (AHA), were included. Films were reviewed, and presence or absence, of dystonia and choreoathetosis were scored by three experienced raters.
Results: Dystonia in the hand was present during activities in 70% (68/97) of the children at a mean age of 12 years (SD 4,4). In 74% (50/68) of these children, dystonia was present more than 50% of the evaluated time. For 63% (43/68) more than one digital recording at younger ages were available. Dystonia could first clearly be observed at a mean age of 3,8 years. Choreoathetosis was observed in 7% (5/68) of the children with dystonia. Children without dystonia had significantly higher (corresponding to better function) AHA units (median: 75, 25th - 75th: 45–82) in comparison to children with dystonia (median: 57, 25th - 75th: 52–63) ($p = 0.01$).
Conclusion: Dystonia in the hand is common in unilateral CP and correlates to lower hand functioning.

1. Introduction

Children with unilateral spastic cerebral palsy (CP) often have considerable difficulties with their hand function. To better understand the obstacles and problems that they meet, we need to identify the different components that contribute to the reduced functioning.

Overall, spasticity and dyskinesia, are considered to be debilitating motor symptoms and are present in individuals with CP in 80–90%, and 5–15% respectively. Accordingly, much focus has been put on reducing or eliminating these. Dyskinetic CP, is characterized by involuntary, uncontrolled, recurring, occasionally stereotyped movements, fluctuating muscle tone, and persistent primitive reflex patterns (SCPE) [1]. In dyskinetic CP two main movement disorders, dystonia and choreoathetosis, often co-exist even though dystonia commonly is the most dominating [2,3].

Contrasting to spasticity, that is identified through a physical examination [4], dyskinesia is visually recognized, and the available rating scales utilize what is observed, often through applying operational criteria to subjective observations through a video recording [3,5]. It is widely recognized that in children with primarily spastic bilateral CP, dystonia can co-occur besides the spasticity and likewise is spasticity often present in individuals with dyskinetic CP(6). The overlapping features of spasticity and dystonia may be hard to differentiate and dystonia may be misclassified as spasticity [7].

In unilateral spastic CP, dystonia of the upper extremity is rarely identified or addressed. The co-occurrence of dyskinesia in spastic CP and the difficulties related to identifying it in the clinical setting, has lately received increased interest [7,8] [6,9-14].

As an example, a 2019 meeting in Banff Canada, the "International Cerebral Palsy Function and Mobility Symposium" concluded that one of the top goals for the upcoming five years was to "develop a better, standardized way of quantifying dystonia and assessing whether it plays a role during walking, to better monitor treatment outcome and quantify patients with primary and secondary symptoms of dystonia" [15].

In unilateral CP, stereotyped upper extremity postures are often identified [7]. These include hypertonic posturing in elbow flexion,

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https://doi.org/10.1016/j.ejpn.2022.06.015
Received 9 January 2022; Received in revised form 2 June 2022; Accepted 26 June 2022
Available online 11 July 2022

forearm pronation, wrist flexion, thumb adduction, and finger flexion. These postures have traditionally been attributed to and labeled "spasticity" but can be caused by other factors such as dystonia [9]. In fact, specific stereotyped postures and movements in dystonia have been described [16]. For the upper extremity these include but are not restricted to; swan neck position, finger hyper extension, the one-two-five position, palmar flexion of the wrist and a wrist drop position. Thus, children with spastic unilateral CP might even have elements of dystonia and identifying this may be important when guiding, improving and choosing interventions.

This trial aimed to better understand if and to what extent dystonia was present during activity, in the upper extremity, of children diagnosed with spastic unilateral CP. Additionally, we aimed to describe how dystonia, if present, developed longitudinally and at what age it was first detectable.

2. Materials and methods

2.1. Study design and study population

This is a descriptive study evaluating the presence of dystonia during hand activities in children with unilateral spastic CP. All children born between 1999 and 2014, with a confirmed diagnosis and classification of unilateral CP at 4 years of age or older [17] and having digital recordings of hand assessments were included. Digital recordings from 2000 to 2019 were screened.

2.2. Assessments and classifications

2.2.1. Videos

At the Neuropediatric unit, the Department of Woman's and Children's Health at Karolinska Institutet, there has been a great emphasis on hand function, assessment tools, and intervention studies in CP. With parental consent, videos from hand assessments, often repeatedly captured from children with primarily spastic unilateral CP were stored and a database constructed. In this project, we utilized the existing videos and data that were collected from assisting hand assessments (AHA).

Ethical approval from the Regional Ethics Committee in Stockholm (2001/00–436 2008/148-31, 2011/278-32, and 2018/1329-32) was attained.

2.2.2. Hand assessment

In research and clinical work, AHA and mini-AHA is commonly used in unilateral CP from the age of 8 months–18 years of age [18,19]. The AHA consists of 20 age-appropriate test activities that describe object related hand actions, scored on a four-point rating scale. The AHA gives sum scores that are converted to AHA-units, ranging from 0 to 100 (100 being the best) [20]. AHA has shown excellent psychometric properties [21]. During an AHA -evaluation the child will always be seated in a stable position in front of a table and be filmed when handling a range of objects according to a standardized method [21]. In this study, we report AHA units, when available, for the children at age 5 years ± 6 month.

2.2.3. Participants

From the available database we collected; sex, Gross Motor Function Classification System (GMFCS), and Manual Ability Classification System (MACS) levels (ranging from 1 to 5, highest functioning in level 1), additionally, the laterality of the affected hand was identified from the films.

2.3. Procedure

2.3.1. Rating of dystonia

We used the definition of dystonia as a movement disorder that is characterized by sustained or intermittent muscle contractions causing abnormal, often repetitive movements, postures, or both [22]. For the upper extremity, these may include the specific stereotyped finger, wrist, and elbow, postures, and movements that are often present in dystonia as described by Sanger et al. [16]. From the digital recordings we observed if we could identify dystonia in CP with characteristic involuntary and/or distorted voluntary movements, and abnormal postures due to sustained muscle contractions in the affected hand during activities, were identified [1]. Specifically, we reviewed the digital recordings for dystonia in the distal arm, including the forearm, the wrist, and the hand. The duration of dystonia during different types of hand activities was scored as absent (0), clearly present during less than 50% of the activities [1], or present during 50% or more of activities [2].

2.3.2. Rating of choreoathetosis

If choreoathetosis of the distal arm was observed, this was noted as present or absent and not quantified. In CP this is dominated by hyperkinesia and hypotonia. The muscle tone is mostly decreased but fluctuating and constantly changing with fragmented or contorting movements [1]. For unspecific, or very brief signs of dystonia and choreoathetosis, or if visible only during one specific activity (such as finger hyperextension only when reaching to grasp an object) we had a "benefit of doubt approach" and rated 0 or no clear dystonia/choreoathetosis.

2.3.3. Reviewing videos

For all children included in the study, a minimum of one available digital recording (film) after four years of age was required. A large proportion of the recordings made during the first part of the period had utilized VHS or CD/DVD type of videos that had not been digitally converted. These videos could not be reviewed.

We reviewed the films and assessed the presence or absence of dystonia, and if present the duration during activity. All films were jointly assessed and screened by all authors, three senior clinicians, and researchers with many decades of experience in all types of CP, and other movement disorders, such as inherited dystonia. For all children, we started with the most recent film, meaning that there were no later available for that specific individual, and the age of the child was documented. This method was chosen since dystonia, in clinical practice, is often considered to develop at a later age than for example spasticity and also to be more easily identified at an older age. If we identified dystonia in the most recent film, we proceeded to evaluate the earliest available film for that child, to identify the youngest age when dystonia was clearly recognized. If a child that had dystonia at an older age did not display dystonia in the first film made at a lower age, we continued reviewing all films chronologically for that child until the dystonia appeared in order to identify the age when dystonia could be first observed.

If dystonia was observed during activity in a child, we ensured that the wrist and hand could be observed in a neutral position, to ensure that reduced range of motion was not the explanation for the abnormal positions and movements. We also confirmed that different types of hand activity/movements such as grip, holding, releasing, reaching, twisting/screwing, pointing, lifting, or pushing was clearly seen in the recording. Even if dystonia was identified during different activities early on in the video, we pursued and watched a minimum of 5 min of each film.

During the process, we identified seven specific recurrent positions when dystonia was present. Some of these positions, "swan neck position", "one-two-five position" and "fisting" had already been identified and named in a study by Sanger et al. [16]. Additionally, we identified three more positions, that were named; "palmar flexion" "finger hyper extension", and "finger spreading", see, Supplementary Fig. 1 for photo illustrations. Palmar flexion was characterized by a palmar flexed position of the wrist, finger hyperextension by hyperextension of at least one of the phalangeal joints, and finger spreading by abduction of at least two of the fingers. Interestingly enough, most children with

dystonia exhibited several of these stereotyped positions, often occurring simultaneously, Supplementary Fig. 1 shows the presence of various dystonic positions in the children.

If a child did not display any dystonia during the latest available digital recording, we did not evaluate earlier digital recordings for that child. Before establishing that a child did not have any dystonia the full length of the digital recording was reviewed.

2.4. Statistical analysis

Statistical analyses were performed with SPSS version 26. Descriptive parametric data are presented with mean and standard deviation (SD), and non-parametric data are presented with median, interquartile range (25th - 75th). Wilcoxon Signed Rank test (two related samples) was used to analyse differences in the dystonia scores [1,2] from the earliest digital recordings where dystonia was clearly identified, with the scores from the latest digital recordings. Mann-Whitney test (two independent samples) was used to analyse differences between AHA scores in children with and without dystonia. Significance level was set at $p < 0.05$. Correlations of non-parametric data were calculated by using Spearman's correlation coefficient (r_s) between absence/presence of dystonia, MACS and AHA scores. Correlations were considered significant when $p < 0.05$ and $r_s > 0.30$. The following interpretation was used for the size of the correlation: r_s: 0.00–0.30 negligible correlation, 0.30–0.50 low correlation, 0.50–0.70 moderate correlation, 0.70–0.90 high correlation, and 0.90–1.00 very high correlation (Hinkle 2003, book chapter).

3. Results

Ninety-seven children born between 1999–2014, with a diagnosis of spastic unilateral CP, and standardized digital films were included (Table 1). During the process, it was clear that a substantial part of children that were filmed before 12 months of age had their affected hand fisted. The hand was often fisted through the major part of the film and with a lack of variation in the movement patterns, thus we could not evaluate activity nor ensure a neutral position of the hand. Consequently, we decided not to include any videos before 12 months of age.

The average film was 20 min (SD 5 min) long. Observations from the last video recordings revealed the presence of dystonia in 70% (68/97) of the children (Table 1). In 74% (50/68) of these children, dystonia was present more than 50% of the evaluated time. In 63% (43/68) of these children, there were earlier available digital recordings. The mean age of 3,8 years (SD 1,5) was identified to be the youngest age when dystonia could clearly be observed. The dystonia scores [1,2] from the earliest digital recordings where dystonia was clearly identified were compared with the scores from the latest digital recordings. In 70% (30/43) of these children the dystonia scores were unchanged, in 26% (11/43) the scores increased, and in 4% (2/43) the dystonia scores decreased at the latest occasion ($p = 0.01$). In addition, the presence of choreoathetosis was observed in 7% (5/68) of the children with dystonia in the most recent films (Table 1).

Children without dystonia had significantly higher (corresponding to better function) AHA units (median: 75, 25th - 75th: 45–82) in comparison to children with dystonia (median: 57, 25th - 75th: 52–63) ($p = 0.01$). This became even more pronounced in comparison between children without dystonia to those with a duration of dystonia more than 50% of the time (median: 55, 25th - 75th: 48–58 ($p = 0.004$). A correlation between the presence of dystonia at the most recent films (the films were the children had the highest available age) and lower AHA units was observed ($r_s = 0.41$, $p < 0.001$). Similarly, there was a correlation between presence of dystonia at the most recent films and MACS ($r_s = 0.47$, $p < 0.001$) with those that did not exhibit dystonia having a better ability level within MACS.

Table 1
Descriptive data of the **Total** group (n = 97), the group with **Presence** of dystonia (n = 68), and the group with **Absence** of dystonia (n = 29).

Descriptives	Total	Presence	Absence
Sex n (%)	**97 (100)**	**68 (100)**	**29 (100)**
Male	50 (52)	37 (54)	13 (45)
Female	47 (48)	31 (46)	16 (55)
Unilateral CP, laterality n (%)	**97 (100)**	**68 (100)**	**29 (100)**
Right	58 (60)	43 (63)	15 (52)
Left	39 (40)	25 (37)	14 (48)
MACS level n (%)	**94 (97)**	**66 (97)**	**28 (97)**
MACS I	31/94 (33)	14/66 (21)	17/28 (61)
MACS II	51/94 (54)	43/66 (65)	8/28 (29)
MACS III	12/94 (13)	9/66 (14)	3/28 (10)
GMFCS level n (%)	**79 (81)**	**55 (81)**	**24 (83)**
GMFCS I	71/79 (90)	50/55 (91)	21/24 (88)
GMFCS II	8/79 (10)	5/55 (9)	3/24 (12)
AHA scores at 5 years n (%)	**88 (91)**	**63 (93)**	**25 (86)**
Median (25th - 75th)	58 (52–70)	57 (52–63)	75 (45–82)
Age when AHA was assessed n (%)	**88 (91)**	**63 (93)**	**25 (86)**
Years, Mean (SD)	5 (0,3)	5 (0,3)	5 (0,4)
Presence/absence of dystonia n (%)	**97 (100)**	**68 (100)**	**29 (100)**
No dystonia (0)	29 (30)	0 (0)	29 (100)
Dystonia <50% of the film (1)	18 (19)	18 (26)	0 (0)
Dystonia >50% of the film (2)	50 (51)	50 (74)	0 (0)
Dystonia total n (%)	68 (70)	68 (100)	0 (0)
Presence of choreoathetosis n (%)	**5 (4)**	**5 (7)**	**0 (0)**
Age at the last video n (%)	**97 (100)**	**68 (100)**	**29 (100)**
Years, Mean (SD)	11,6 (4,2)	12 (4,4)	10,6 (3,5)
Video length (the last) n (%)	**97 (100)**	**68 (100)**	**29 (100)**
Minutes, Mean (SD)	20 (5)	20,1 (6,0)	18,8 (5,1)
First time dystonia was observed[a] n (%)		**43 (63)**	
Dystonia <50% of the film (1)		21 (49)	
Dystonia >50% of the film (2)		22 (51)	
Age the first time dystonia was observed n (%)		**43 (63)**	
Years, Mean (SD)		3,8 (1,5)	
Video length (the first) n (%)		**43 (63)**	
Minutes, Mean (SD)		18,5 (4,7)	

Data are presented as number (n), percentage (%), mean, standard deviation (SD), median and percentile 25 and 75 (25th - 25th). Cerebral palsy (CP), Manual Ability Classification System (MACS), Gross Motor Function. Classification System (GMFCS), and Assisting Hand Assessment (AHA). At the latest available digital film 68 children had dystonia, 43 of these children had earlier digital films.

[a] Only children with dystonia observed at the last film.

4. Discussion

In this study we identified clear dystonia in 70% of all children with spastic unilateral CP. For those children where we could follow hand activity longitudinally from 12 months and onwards, dystonia was first clearly observed at 3 years and 10 months of age. When we compared the extent of dystonia in the first available digital recording, to the most recent film where dystonia was present in a specific child, most children had unchanged amount of dystonia, while 26% showed increased dystonia and 4% showed decreased dystonia at later age.

Even though, the overall proportion of children with unilateral CP that did exhibit dystonia in their upper extremity during activity surprised us, it is well in line with a previous report [10]. In an Australian study, 151 children, 85% classified with spastic CP, were evaluated for dystonia through a broad protocol [10]. The study found that an astonishing ~80% and ~60% of children having primarily spastic type CP had identifiable dystonia of the lower limbs, and upper limbs respectively [10].

In our study, children with dystonia had more difficulties to perform bimanual activities and displayed lower AHA scores in comparison to

those without dystonia. Similar findings have been reported in a study including 100 children with CP, ages 3–18 years, that reported lower hand ability in children with dystonic and athetoid CP, in comparison to children with spastic CP [23]. In addition, our results revealed that the presence of dystonia to some extent correlated with MACS levels and lower hand ability as assessed with AHA. This is in accordance with results from a study that found high correlations between dystonia severity and MACS levels and dystonia and GMFCS levels in children with dyskinetic CP [10].

Many children with CP have a mix of different motor and other neurological symptoms. Therefore, for each individual, the recommendation of classifying the CP subtype with respect to the most dominating symptom is not uncomplicated [24]. It has been recommended that the presence of each of the tone and/or movement abnormalities in fact should be identified, and listed, and that this approach may be of greater clinical and etiologic utility compared to when only addressing the most dominating symptom [6]. Consequently, using assessment tools to identify both dystonia and spasticity in individuals diagnosed as having either spastic CP or dyskinetic CP is of importance in the clinical setting, for prognosis and when planning for interventions [13]. Although, the distinction between dystonia and spasticity is difficult to ascertain, simply being aware of the risk of co-occurrence will be helpful by extending the clinical examination to include assessments that cover both symptoms [13]. Whether the hand positions typically observed in unilateral CP are attributed to spasticity or dystonia, or a combination of the two disturbances, may have consequences for choosing interventions and the subsequent outcome. Supporting this are the results from large comprehensive systematic reviews where it is apparent that those interventions that effectively reduces muscle tone does not, to the same extent, improve motor function nor the improvement of tasks [11,12]. Muscle tone interventions are mainly focusing on spasticity while "less research contains dystonia management, given the lower prevalence and under-recognition of this motor disorder" which could effectively contribute to the less than anticipated effect that has been observed after certain spasticity reducing interventions [12]. A study in the Netherlands established that 9 of 24 (38%) non-walking patients with spasticity exhibited some signs of dystonia following selective dorsal rhizotomy [25]. Not surprisingly, when spasticity was reduced and the non-previously identified dystonia became obvious, the caregivers were more likely to be unsatisfied with the procedure [25].

Furthermore, identifying dystonia in children with unilateral CP could possibly guide orthopeadic management. In dyskinetic CP, and other childhood dystonia, musculoskeletal deformities are often associated with pain, therefore orthopaedic surgery might be a recommended treatment. Based on clinical experience, the results of soft tissue orthopaedic surgery in patients with dyskinetic CP are not as predictable as in patients with spastic CP, and adverse outcomes can easily occur [10,26].

Purely observing a child during movement and play in various activities with the awareness of the stereotyped positions and movements described for dystonia such as; palmar flexion, finger hyper extension and fisting, in mind, may be helpful to identify the presence of dyskinesia. In our study, in the same child, the dystonic positions and movements often co-existed in various combinations. This might imply that any observation aiming to identify dystonia should be sufficiently long. The AHA assessment is developed to measure how effectively the involved hand is used for bimanual activity, and AHA can be used to follow development over time since the test is sensitive to change [21]. AHA is not developed to provide information on what disorders may be associated with the difficulties. All assessments in the present study, were filmed, and scored after testing. Therefore, combining assessments of hand function with observations of dyskinesia using for example available AHA-films and well-defined descriptions of the phenomena might be a first step to a more standardized screening for dystonia in CP.

In our study, dystonia was first observed at a mean age of close to four years of age. Preferably any screening for dystonia could be performed at a slightly later age to ensure classification of CP subtype and possible coexisting secondary symptoms or at an earlier age if needed to choose an intervention. If dystonic movements or postures are suspected, further investigations with for example the dyskinesia impairment scale (DIS) could be used to confirm the results [3].

When the children were less than 1 year of age, we observed that they commonly kept the affected hand in a constantly fisted position with sparse variation in the movement pattern and, as a result, almost no involuntary movements were observed. At this young age, we did not consider the fisting as a sign of dystonia since it could not be differentiated from either spasticity or limited range of motion, especially since no physical examination was available. At an older age the hand opened up, and dystonic movements and positions were often observed. Not unlikely, the fisted hand to some extent masks dystonia in a child with unilateral CP at younger ages. This observation requires to be further investigated since dystonia commonly is considered a neurological symptom that is developed over time in CP. Furthermore, the dystonia scores remained unchanged over time from when first observed, at younger ages, compared to when last assessed, at around 11 years of age, in most children. Even so, it must be noted that severity of dystonia increased in about 25% of the children and age must be considered when assessing for dystonia.

Dystonia has been proposed to be visually assessed through observations during movements or at rest [3]. We screened for dystonia during activities only. We did not include dystonia during rest since many children had their hands in their lap while resting, obscured by the table, making a consistent evaluation impossible. In a study on clinical patterns of dystonia in 55 individuals with dyskinetic CP assessed with the DIS, both dystonia and choreoathetosis increased during activity compared to at rest [2].

4.1. *Strengths and limitations*

There were several strengths and limitations in the present study. A strength was that all children were assessed in a structured and standardized approach by AHA examinations. The AHA assessments were recorded and therefore offered the opportunity to carefully observe and assess the absence or presence of dystonia in different activities. A further strength was the number of children included and the longitudinal evaluations of their development. In addition, the assessments were performed by three senior clinicians, and researchers, with many decades of experience in all types of CP, and other movement disorders, such as inherited dystonia.

Presence of dystonia were assessed by observations of films without a physical examination, which can be considered as a limitation, however, dystonia has been proposed to be visually assessed through observations during movements or at rest [3]. Digital recordings were not available for some of the older children when they were young, implying that we could not assess their films. Another limitation was the identified age when dystonia could first be observed, this was dependent on the time intervals between films, which means that dystonia could possibly occur earlier than what was captured on film at a specific time.

5. Conclusion

In a summary; This study identified dystonia in the upper extremity in 70% of children with spastic unilateral cerebral palsy when recordings of structured bimanual activities were reviewed. A correlation was seen between the presence of dystonia and lower hand ability when MACS classification, and AHA scores at 5 years of age was utilized to quantify functioning. This implies that identifying dystonia in spastic unilateral cerebral palsy is important. Additionally, this study identified a range of different stereotyped positions that were existing in the hand and wrist, when dystonia was present that requires careful evaluations.

Declaration of competing interest

All authors have agreed to the final version of the manuscript. There are no conflicts of interest and no sponsor have been involved in any part of this research.

Acknowledgements

We want to thank the children, the parents and the therapists who made this study possible. We also want to thank Professor Ann-Christine Eliasson, for her thorough and long-term commitment that resulted in this study.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.ejpn.2022.06.015.

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